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Abstract: Moyamoya vasculopathy (MMV) leads to chronic hypoperfusion predominantly in the middle cerebral artery (MCA) and anterior cerebral artery (ACA) territories. Most revascularization techniques focus on revascularization of the MCA territory. Augmentation of blood flow in the frontal area is important for neurocognition and lower extremity function. In this article we describe a new combined (direct and indirect) one-stage bypass technique consisting of a superficial temporal artery to middle cerebral artery (STA-MCA) bypass with encephalo-duro-synangiosis (EDS) for unilateral MCA revascularization, along with an encephalo-duro-periosteal-synangiosis (EDPS) for bifrontal blood flow augmentation. The strength of this technique is the revascularization of three vascular territories during a single surgical intervention: the MCA unilaterally; and the frontal territories bilaterally. Bifrontal EDPS may also be considered as a supplementary independent procedure for patients who previously underwent revascularization treatment in the MCA territory, but develop symptoms due to frontal hypoperfusion.

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SUMMARY

Moyamoya vasculopathy (MMV) leads to chronic hypoperfusion predominantly in the middle cerebral artery (MCA) and anterior cerebral artery (ACA) territories. Most revascularization techniques focus on revascularization of the MCA territory. Augmentation of blood flow in the frontal area is important for neurocognition and lower extremity function. In this article we describe a new combined (direct and indirect) one-stage bypass technique consisting of a superficial temporal artery to middle cerebral artery (STA-MCA) bypass with an encephalo-duro-myo-synangiosis (EDMS) for unilateral MCA revascularization, along with a encephalo-duro-periosteal-synangiosis (EDPS) for bifrontal blood flow augmentation. The strength of this technique is the revascularization of 3 vascular territories during a single surgical intervention: the MCA unilaterally and the frontal territories bilaterally. Bifrontal EDPS can also be considered as a supplementary independent procedure for patients who previously underwent revascularization treatment in the MCA territory, but develop symptoms due to frontal hypoperfusion.

Key words: bilateral frontal territories; bypass surgery; STA-MCA bypass; indirect revascularization; moyamoya; neurocognition.

Abbreviations:

ACA: anterior cerebral artery; CBF: cerebral blood flow; CVR: cerebrovascular reserve; EDS: encephalo-duro-synangiosis; EDMS: encephalo-duro-myo-synangiosis; EMS: encephalo-myo-synangiosis; EDPS: encephalo-duro-periosteal-synangiosis; EPS: encephalo-periosteal-synangiosis; H_2O-PET: H_2O-positron emission tomography; ICA: internal carotid artery; IF: interhemispheric fissure; MCA: middle cerebral artery; MMS: moyamoya syndrome; MMV: moyamoya vasculopathy; SSS: superior sagittal sinus; STA: superficial temporal artery; TIA: transient ischemic attack.

INTRODUCTION

Moyamoya vasculopathy (MMV) is a rare cerebrovascular disease of unknown etiology that is characterized by progressive bilateral stenosis of the intracranial internal carotid arteries (ICAs) and their proximal branches, resulting in cerebral hypoperfusion with subsequent ischemic symptoms or, less often, intracerebral hemorrhage. If MMV is associated with other disease conditions (i.e.: Down's syndrome, neurofibromatosis type I), patients are diagnosed with moyamoya syndrome (MMS). [21]

In MMV, reduced perfusion in the anterior circulation leads to compensatory development of collateral vasculature by small vessels near the apex of the carotid, on the cortical surface, leptomeninges, and branches of the external carotid artery supplying the dura and the skull base. Rarely do these processes involve the posterior circulation. [22] Surgical cerebral revascularization is considered the only effective treatment modality and can be achieved by direct, indirect or combined methods. [1,3,14] Direct techniques consist of an anastomosis of a donor artery, generally the superficial temporal artery (STA), to a cortical recipient arterial branch of the middle cerebral artery (MCA), which instantly augments blood supply. Indirect methods for cerebral revascularization are based on the approximation of vascularized tissue such as the temporal muscle, pericranium, dura or omentum onto the cortex in order to promote neoangiogenesis over time. [1, 4] Combined techniques provide the advantages of both methods, however there is no evidence of one technique having advantages over the other. [3] Most techniques focus on revascularization of the MCA territory. However, revascularization in the frontal territory is receiving gradually more attention: a considerable part of the ischemic presentation of (pediatric) moyamoya patients consists of neurocognitive disorders as well as lower extremity function, caused by frontal hypoperfusion. [2,5,12,24] Re-establishing cerebral blood flow (CBF) in the frontal territory may prevent, stabilize or improve neurocognitive decline. [6-7,15,17-20,24]

In this paper, we describe a combined one-staged procedure for revascularization of the MCA territory unilaterally by a direct STA-MCA bypass and the bifrontal territory by an encephalo-duro-periosteal-synangiosis (EDPS). To illustrate the procedure, the treatment of a girl affected by MMS is reported.

MATERIALS AND METHODS

Illustrative case

A 9-year old girl affected by neurofibromatosis type I presented with multiple transient ischemic attacks (TIA's) consisting either of diplegia of the legs or monoparesis of the left arm, combined with frequent headaches. Neurological examination showed no deficits. MMV was documented on magnetic resonance imaging, angiography and digital subtraction angiography, demonstrating bilateral occlusion of the terminal ICA and the M1 and A1 segments, with bilateral typical compensatory moyamoya vessels.The patient was diagnosed as having MMS. H_2O-positron emission tomography (H_2O-PET) scan documented extensive decreased baseline

CBF in the right MCA and frontal area as well as severely impaired cerebrovascular reserve (CVR) in almost the entire right hemisphere after the administration of acetazolamide. Moderately decreased baseline CBF was seen in the left hemisphere, especially the frontal area, with well-preserved CVR in the MCA and posterior cerebral artery territory, but decreased frontal CVR (Figure 1 A, B). Based on the symptomatic course of the disease and the hemodynamic insufficiency, surgical revascularization was indicated and aimed at the augmentation of blood supply to the right MCA and both frontal territories.

Combined technique for unilateral MCA and bifrontal revascularizazion

After general anesthesia, the patient was placed in supine position with the head mildly extended and rotated 30° to the left side in a Mayfield headrest. The first procedure step consisted of a right-sided STA-MCA bypass along with an encephalo-duro-myo-synangiosis (EDMS). A linear incision located behind the hairline over the course of the STA was performed, without hair shaving. The classic technique of a STA-MCA bypass was followed. [9] The parietal branch of the STA was dissected and the small arising branches coagulated and cut. The donor vessel was kept intact until the anastomotic procedure was started. The temporal muscle was cut creating a flap with its base directed towards the skull base, to perform an encephalo-myo-synangiosis (EMS) at a later stage. A craniotomy was performed along the Sylvian fissure. The dura mater was thereafter carefully opened in a star fashion to maintain the main branch of the middle meningeal artery and dural flaps were reflected over the brain surface underneath the bone window: encephalo-duro-synangiosis (EDS) was obtained by creating contact between the external surface of the dura and the brain surface. Following, the cortex was surveyed for the largest cortical recipient artery. After identification of the most suitable recipient vessel (M4 branch > 0.8 mm in diameter without microperforating vessels), it was dissected by means of arachnoid opening. A temporary non-traumatic microvascular clip was placed across the proximal exposed STA. A fish mouth cut was then executed at the distal STA to increase the opening diameter of the donor vessel. A blue dye was put onto the surface of the cutting edges of the donor and recipient vessels to visualize the edges clearly during the anastomotic procedure. Non-traumatic microvascular clips were applied on the recipient vessel and two silicon triangle-shaped background sheets inserted beneath, in order to simplify the construction of the anastomosis. Thereafter, a linear arteriotomy on the cortical recipient vessel was performed on the blue dyed line. Two 10-0 monofilament sutures were used to anchor the donor and recipient vessel at the toe and the heel of the anastomotic site. The anastomosis was then completed by means of interrupted microsutures, to allow eventual anastomosis growth with time. First the distal and then the proximal temporary clips on the cortical MCA recipient artery, and subsequently the clip on the proximal STA were removed. Verification of the bypass patency was performed by Indocyanine Green Videoangiography (performed by using a commercially available surgical microscope, OPMI® Pentero™, The Carl Zeiss Co, Oberkochen, Germany). Bidirectional flow through the MCA was assessed and all flows were quantified (18 ml/min) using a flow probe (Transonic system, Inc.© Ithaka, NY, USA). After establishing the bypass, EMS performed by covering the exposed cortex with the temporal muscle and by suturing the muscle to the dural edges: attention was paid to avoid compression of the bypass. The previously performed burr hole was enlarged to permit the intracranial entrance of the bypass without risk of compression. The bone was then returned to its normal position and secured in place.

Bifrontal revascularization through EDPS was performed by extending the skin incision 4 cm over the midline in a zig-zag fashion to optimize cosmetic results. The scalp flap was reflected anteriorly and a vascularized pediculated (towards the bitemporal and biorbital regions) bifrontal pericranial flap consisting of periosteum was prepared by careful dissection (Figure 2A, Figure 3A) After completion of two separate frontal parasagittal craniotomies (dimensions 4 x 5 cm) (Figure 2A, Figure 3B), the dura was opened in a star-shaped fashion and a bifrontal EDS was performed by inverting the dural flaps beneath the edges of each bone window (Figure 2A). The craniotomies were localized 2 cm away from the midline to avoid injuring the superior sagittal sinus (SSS) and the parasagittal veins. Hereafter, the bifrontal encephalo-periosteal-synangiosis (EPS) was accomplished by small arachnoidal openings with positioning of the previously prepared pediculated periosteal flap over the cortical convexity on both sides (Figure 2B). Finally the periosteal flap was sutured laterally to the dural edges, the two frontal bone opercula repositioned and fixed and the scalp flap re-approximated and closed. (Figures 2C, Figure 3C).

RESULTS

The patient had no new neurological deficits after awakening in the pediatric intensive care unit. After hospital dismission, she only experienced 3 short episodes with diplegia and a monoparesesis of the left arm, that were diagnosed as TIA's, in the first 3 months following surgery, but never had recurrent ischemic symptoms in the almost 2 years that followed. Furthermore, a good cosmetic outcome was objectified. The one-year and 8-months post-operative H_2O-PET documented clear improvement of CBF and CVR in the right hemisphere and the left frontal regions. The non-operated left MCA territory showed decreased CVR compared to preoperatively. (Figure 1C-D)

DISCUSSION

Next to revascularization in the MCA territory, re-establishing CBF in the anterior cerebral artery (ACA) area is most likely to be of importance, in particular in children with MMV[20,23-24]; neurocognitive development has been correlated to frontal lobe CBF. [18, 24] In this report we described a cerebral revascularization technique for patients with MMV which enables increasing CBF in three different vascular territories. The described procedure permits to: 1) achieve an immediate unilateral MCA flow augmentation by performing a STA-MCA bypass and reinforce the blood supply by promoting neoangiogenesis in the MCA territory by means of EDMS, and 2) revascularize the ACA territories bilaterally by means of EDPS.

Revascularization with frontal pericranial flaps in patients with MMV has been shown to be effective in promoting frontal synangiosis. [20] In the technical modification presented, the revascularized frontal area is further expanded by inverting and reflecting dural flaps over the cortex for each craniotomy. This bifrontal EDPS method is easy to perform and can be applied in patients with MMV who suffer from bifrontal hypoperfusion with hemodynamic impairment documented on PET examination. It represents a good alternative to the existing techniques for frontal territories revascularization such as the technically challenging direct STA-ACA bypass or other procedures that need exposure of the interhemispheric fissure (IF). [10-13]. In fact, the use of separate frontal parasagittal craniotomies, located one on the left- and one the right-side 2 cm away from the midline, reduces the risk of injuries to the SSS and the parasagittal veins. With this technique, no exposure of the IF is needed. Furthermore, the procedure does not compromise eventual future contralateral MCA territory revascularization. By placing the incision behind the hairline in a zig-zag fashion with a non-shaving policy, a nice cosmetic result can be ensured.

It is also important to note that bifrontal EDPS itself could be used as a supplementary procedure, even in patients who already underwent other cerebral revascularization procedures (typically aimed to augment blood supply in the MCA territory), in case they would develop symptoms related to hypoperfusion of the bifrontal territory or in case of progression of MMV.

This technique represents a modification of previously reported techniques for the revascularization of the frontal lobes. (8, 11,13, 16, 18, 20, 23) The novelty of this technique is unfolded in the one-staged approach combining direct and indirect revascularization in three different areas, with minimal surgical risks (by avoiding the IF and SSS). To our knowledge, this one-staged procedure has not been reported in the literature.

CONCLUSIONS

The one-staged technique for contemporary revascularization of 3 different vascular territories as described here allows unilateral combined revascularization of the MCA territory by means of direct STA-MCA bypass and EDMS, as well as indirect revascularization of both ACA territories through bifrontal EDPS. This technique seems to be a safe and effective alternative treatment for patients with hemodynamic compromise in both frontal territories in addition to the MCA region. Furthermore, the EDPS itself could be used as a supplementary effective and safe revascularizing option in patients with MMV who already underwent direct or indirect bypass procedures in the MCA territory.

FIGURES AND FIGURE LEGENDS

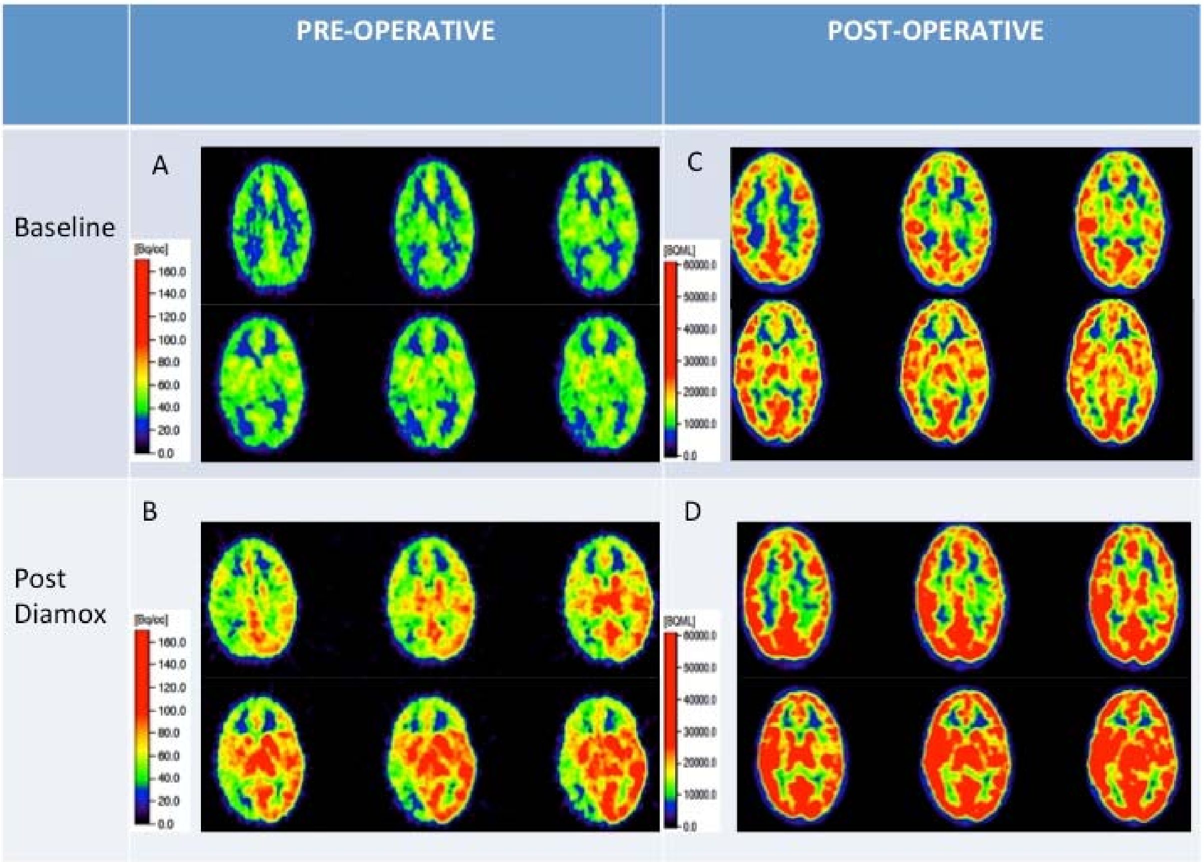


Figure 1

(A,B) Pre-operative H_2O-PET scan revealed severe baseline perfusion deficits and almost absent CVR after acetazolamide in almost the entire right hemisphere. Left hemispheric baseline CBF was moderately affected and CVR was largely preserved, with exception of the left frontal region.

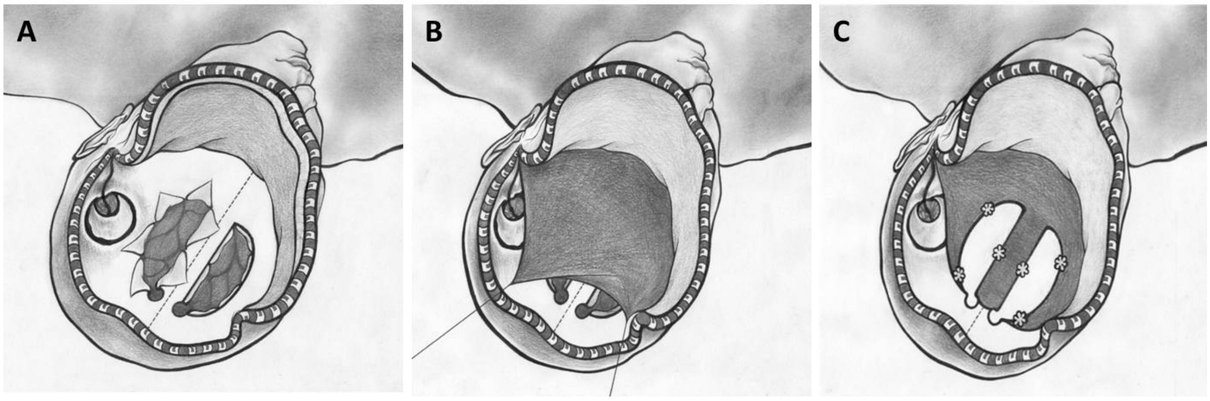
(C, D) One –year and 8 months postoperatively the H_2O-PET revealed clear improvement of CVR in the right hemisphere and the left frontal regions. The non-operated left MCA territory showed decreased CVR compared to preoperatively.

Figure 2: Illustrations of the one-staged technique for revascularization of the left MCA and bifrontal territories.

(A) After performing a STA-MCA bypass (black arrow), the skin incision is extended for 4-cm over the midline behind the hairline, then the scalp flap is reflected anteriorly. The vascularized frontal cranial periosteum is dissected and reflected on the scalp flap (see white asterisk), two symmetric bilateral frontal parasagittal craniotomies are performed and the underlying frontal dura is opened in a star-fashion (white arrow) and inverted on the cortex around each frontal bone window (EDS) (grey arrow).

(B) The dissected pericranial flap (white asterisk) is placed over the cortex (EPS) and sutured to the dural edges.

(C) Finally, the bone flaps are repositioned (black asterisks).



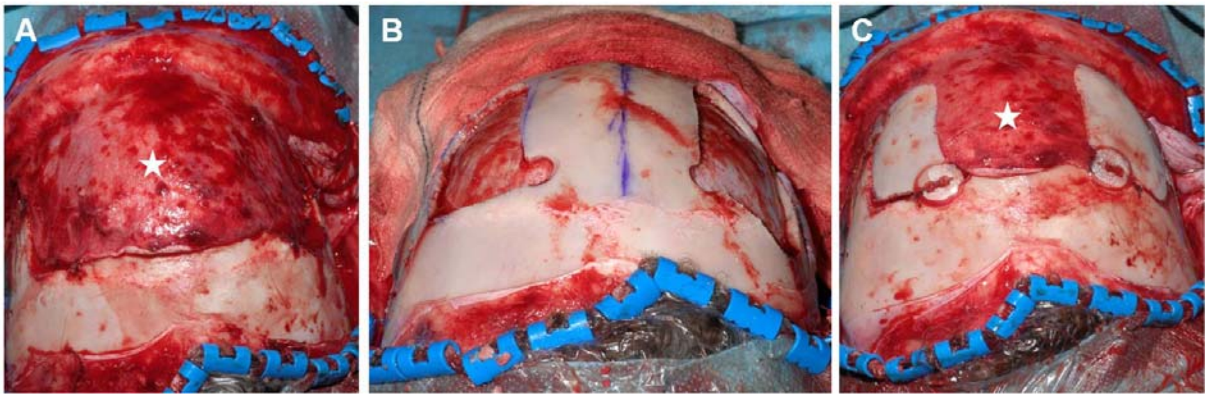


Figure 3: Intraoperative pictures of an EDPS

(A) Dissection and preparation of the vascularized frontal cranial periosteum (white asterisk).

(B) Bifrontal craniotomies located 2 cm from the midline (blue drawn line).

(C) After dural opening in a star-fashion and dura-synangiosis, the periosteal flap (white asterisk) is placed over the cortex, sutured to the dural edges (EDPS), with repositioning of the bone flap.

Disclosure

The authors report no personal financial or institutional interest in any of the drugs, materials, or devices mentioned in this article.

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